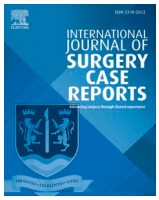
International Journal of Surgery Case Reports 103 (2023) 107860

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report

Atypical presentation of preeclampsia. Case report

Chadia Khalloufi *, Imane Joudar, Mohamed Jalal, Amine Lamrissi, Said Bouhya

Maternity, University Hospital Abderrahim Harouchi, Casablanca, Morocco
Faculty of Medicine and Pharmacy, University Hassan II, Ain chock BP 5366, 20000 Casablanca, Morocco

ARTICLE INFO

Keywords:
Atypical preeclampsia
Clinical manifestations
Screening
Case report

ABSTRACT

Introduction and importance: Atypical pre-eclampsia cases are those that develop before 20 weeks of gestation and after 48 h after delivery and or have some of the signs and symptoms of preeclampsia without the usual hypertension or proteinuria which make them difficult to diagnose.
Case presentation: Our aim is to report a case of atypical preeclampsia (before week 20 of gestation) associated with a HELLP syndrome and analyze the clinical features of atypical forms, assess differential diagnosis and highlight the progress in biochemical and biophysical markers that may help with diagnosis.
Clinical discussion: Severe early pre-eclampsia (before 32 weeks of pregnancy) is associated with a risk of maternal mortality 20 times higher than after 37 weeks, and a higher risk of perinatal complications. Its symptoms are variable and reflect multisystem dysfunction.
Conclusion: Despite the refinement of diagnostic tools available to clinicians, there are still clinical presentations that fall outside the definitions. Any good clinician must be aware of the atypical forms in order to initiate correct management without delay and thus avoid increasing maternal and perinatal morbidity and mortality.

1. Introduction

3 to 5 % of pregnancies results in pre-eclampsia, which remains one of the main causes of maternal-fetal mortality and morbidity worldwide [1]. Lack of prenatal visits due to a series of socio-cultural factors in Morocco such as the higher level of illiteracy in rural landlocked areas with difficult health care access, may increase the risk of preeclampsia which is responsible for 10 to 15 % of maternal deaths in developed countries [2,3].

In general, most women will have a classic presentation of pre-eclampsia (hypertension and proteinuria) after 20 weeks of gestation and/or before 48 h after delivery. However, recent studies have suggested that some women will experience preeclampsia without one or more of these classic findings and/or outside of these time periods. Atypical cases are those that develop before 20 weeks of gestation and after 48 h after delivery and/or have some of the signs and symptoms of preeclampsia without the usual hypertension or proteinuria [4].

Early severe pre-eclampsia (before 32 weeks of gestation) is associated with a risk of maternal mortality 20 times higher than after 37 weeks, and with a higher risk of perinatal complications such as prematurity, fetal growth restriction, premature abruption of the normally inserted placenta and perinatal mortality [5]. Its pathophysiology is complex and multifactorial, with various factors such as genetic, environmental, immunological, and nutritional factors contributing to its occurrence, though the precise cause is largely unknown [6]. The identification of biochemical and biophysical markers that point towards placental and endothelial dysfunction allows us to improve our practices through new screening tests [7], which makes it possible to target pregnancies at risk, and to initiate treatment and early prevention. Currently, the coexistence of arterial hypertension, proteinuria and edema is arbitrary and inconsistent. Preeclampsia can begin in the absence of the clinical data mentioned above or appear before the second half of pregnancy. Its symptoms are variable and reflect multisystemic dysfunctions [5]. Its evolution is unpredictable and can be fulminant. The objective of this article is to report a case of atypical preeclampsia (before week 20 of gestation) associated with a HELLP Syndrome hospitalized in the university hospital Ibn Rochd of Casablanca; and to analyze the clinical characteristics of the atypical forms, the differential diagnosis and the progress in biochemical markers and biophysics that can help in the diagnosis. We ensure that the work has been reported in line with the SCARE 2020 criteria [8].

* Corresponding author at: Maternity, University hospital Abderrahim Harouchi, 19, Tarik Ibnou Ziad Road, 20000 Casablanca, Morocco.
E-mail address: khalloufi.chadia@icloud.com (C. Khalloufi).

https://doi.org/10.1016/j.ijscr.2022.107860
Received 17 November 2022; Received in revised form 23 December 2022; Accepted 29 December 2022
Available online 31 December 2022

2. Case presentation

This is a 35 year old patient, having had three in-utero fetal deaths at five months of pregnancy and one child by vaginal birth (11 years ago), with no particular pathological history, admitted for preeclampsia at 19 weeks of gestation and 4 days, with a systolic blood pressure at 170 mmHg and a diastolic blood pressure of 110 mmHg, headache and epigastric pain. She was brought in by ambulance from her home. On clinical examination, a closed cervix, without detectable bleeding. The fetal heart rate was 145 beats per minute and the obstetric ultrasound was normal. An emergency blood work was carried out, with the result of hemoglobin at 14 g/dL, 142,000 platelets per mm^3, Aspartate-Amino-Transférase (ASAT) at 1550UI/L and Alanine-Amino-Transférase (ALAT) at 938UI/L, lactate dehydrogenase (LDH) at 2251 U/L, 5.2 mg/L of creatinine, and a total bilirubin of 5.21, with proteinuria: urinary protein-creatinine ratio was increased at 600 mg/g. The patient was put on anti-hypertensive treatment and Magnesium sulphate. Faced with the instability of the blood pressure figures and the incomplete HELLP syndrom, a caesarean section was performed by the assistant professor in charge of the case and the patient was transferred for monitoring to the intensive care unit. The fetus delivered was a male weighing 290 g. The evolution was marked by the appearance of a thrombocytopenia at 61,000/mm. After stabilization of her blood pressure under methyldopa and nicardipine and the improvement of the thrombocytopenia to 119,000/mm^3 after a course of corticosteroid therapy, the patient was transferred to the postpartum department and declared discharged on the fifth day with a balanced blood pressure, (systolic: 130 mmHg and diastolic: 80 mmHg) under methyldopa 500 mg/day. Antinuclear antibodies, circulating anticoagulants and anticardiolipin antibodies were negative. The anti and pro-angiogenic factors could not be performed due to lack of means. The urinary protein-creatinine ratio was increased at 600 mg/g.

A follow up of 12 months was ensured, with a physical exam done every month for the first 3 months then every 3 months for the following year, showing a normal blood pressure and a normalization of all her laboratory work within the first month.

3. Discussion

The management is identical to that of classic pre-eclampsia where the choice of therapy (antihypertensive, magnesium sulphate) and the decision to deliver are guided by the presence or absence of signs of gravity [9]. When this syndrome occurs before the 20th week, it is often associated with complete or partial hydatidiform mole and triploidy. Very rare cases of pre-eclampsia before 20 weeks of gestation have been reported in the literature in progressive pregnancies without molar degeneration of the placenta [9]. It has also been described in patients with antiphospholipid syndrome and other cases associated with HELLP Syndrome [10]. A risk factor also found was oocyte donation. In our patient's case, pre-eclampsia was diagnosed at 19 weeks of gestation and 4 days. No molar degeneration was documented. In order to eliminate differential diagnoses that could mimic severe preeclampsia, an antiphospholipid workup was requested in view of the history of unexplained fetal death in utero, which came back negative. The autoimmune workup also ruled out systemic lupus erythematosus. The clinical and biological signs were also not in favour of thrombotic thrombocytopenic purpura or haemolytic uraemic syndrome (renal function not impaired). Once the diagnosis of atypical pre-eclampsia has been made, the pregnancy should be terminated rapidly, once the patient has been stabilized. Magnesium sulphate is given to prevent or control a convulsion. As in our patient's case, the clinical and biological parameters improved once the placenta was removed on the 3rd day postpartum. New early detection strategies are being developed to better target patients at risk. Uterine artery Doppler (UAD) is a non-invasive test that records blood flow in the uterine arteries. In patients who will develop pre-eclampsia, several studies have shown an increase in the resistance index and the pulsatility index as well as the presence of a proto-diastolic notch in the 2nd trimester of pregnancy. Some authors have more recently focused on the UAD in the 1st trimester, as the aim is to introduce preventive aspirin therapy in high risk patients, which is more likely to be effective if started at the end of the 1st trimester. Thus, according to a study of a cohort of 6015 patients at undetermined risk of pre-eclampsia [11,12], the mean pulsatility index (PI) adjusted for gestational age, ethnicity and body mass index, has a detection rate of 41.1 % for pre-eclampsia and 81.8 % for early pre-eclampsia, with a false positive rate of 10 %. The predictive validity of clinical markers alone is lower than that of the combination of clinical markers and the velocimetric indices of the DAU. As for the use of maternal biochemical markers as a screening tool for pre-eclampsia, we cite PAPP-A (Pregnancy-associated plasma protein-A) which plays an important role in the local proliferative response, including trophoblastic invasion [13]. Decreased PAPP-A levels in the first trimester are associated with an increased risk of pre-eclampsia. However, the proportion of subjects with pre-eclampsia who have a PAPP-A concentration below the 5th percentile is only 8–23 % [14], which is insufficient for PAPP-A to be used as an isolated screening test. Recently, several models of combined screening, combining clinical, biochemical and biophysical data (DAU), have been reported. Thus, for the screening of early preeclampsia in the 1st trimester of pregnancy, the following have been combined:

- Mean uterine artery PI, serum PlGF level, mean arterial pressure, BMI, ethnicity, family history of preeclampsia, parity and personal history of preeclampsia [11].
- Serum PAPP-A, PP-13 and mean uterine artery IP [15].
- Lowest uterine artery PI, ethnicity, chronic hypertension, parity and mode of conception and mean arterial pressure [16,17].
- Mean uterine artery PI, serum PAPP-A level, parity and history of pre-eclampsia [12].

However, these different models have not been compared with each other, and the procedure for choosing the best predictive model in the different studies is not always explicit. We also mention in the context of the evaluation of angiogenic markers, the calculation of the fms-like tirosina cinasa-1/placental growth factor index, has also been described as useful in the diagnosis of early preeclampsia and HELLP Syndrome, with a sensitivity of 92 % [10].

The primary take-away lesson is to be able to recognize the early signs of the atypical forms of preeclampsia in order to initiate correct management without delay and avoid complications.

4. Conclusion

Pre-eclampsia is a syndrome with a range of non-specific signs and symptoms and multiple possible etiologies. Despite the refinement of diagnostic tools available to clinicians, there are still clinical presentations that fall outside the definitions. Any good clinician must be aware of the atypical forms in order to initiate correct management without delay and thus avoid increasing maternal and perinatal morbidity and mortality. Biophysical and biochemical markers are currently being evaluated to provide the clinician with an effective tool to identify patients with atypical presentations at an early stage.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

The authors institute provided ethical approval for this case study.

Sources of funding

Not applicable, we declare that we didn't require any funding.

Author contribution

Chadia Khalloufi: Conceptualization, data curation, writing original draft preparation, reviewing and editing.
Imane Joudar: Writing, reviewing and editing.
Mohammed Jalal: Supervision.
Amine Lamrissi: Supervision.
Said Bouhya: Validation.

Guarantor

Khalloufi Chadia.

Research registration number

Not applicable.

Declaration of competing interest

The authors declare that they have no conflict of interest.

References

[1] C. Ronsmans, W.J. Graham, Maternal mortality: who, when, where, and why, Lancet 368 (2006) 1189–1200.
[2] P. Dasari, S. Habeebullah, Maternal mortality due to hypertensive disorders of pregnancy in a tertiary care center in southern India, Int. J. Gynaecol. Obstet. 110 (2010) 271–273.
[3] S.O. Onuh, A.O. Aisien, Maternal and fetal outcome in eclamptic patients in Benin City,Nigeria, J. Obstet. Gynaecol. 24 (2004) 765–768.
[4] B.M. Sibai, C.L. Stella, Diagnosis and management of atypical preeclampsia-eclampsia, Am. J. Obstet. Gynecol. 200 (5) (2009) 481.e1–481.e7.
[5] J.F. Romero-Arauz, C. Carranco-Salinas, A. Leaños Miranda, O.A. Martínez-Rodríguez, Preeclampsia atípica. Reporte de un caso, Ginecol. Obstet. Mex. 82 (2014) 354–360.
[6] F.F. Khidri, Y.M. Waryah, F.K. Ali, H. Shaikh, I.D. Ujjan, A.M. Waryah, MTHFR and F5 genetic variations have association with preeclampsia in Pakistani patients: a case control study, BMC Med.Genet. 20 (1) (2019 Dec) 1–2.
[7] Maryne Talon, Physiopathologie de la pré- éclampsie : état des lieux des connaissances actuelles: étude bibliographique, Gynécol. Obstét. dumas-01464945f vol 1 (2016) 10–37.
[8] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[9] Agnès Ditisheim, Michel Boulvain, Olivier P. Irion, Antoinette Pechère-Bertschi1, Rev. Med. Suisse 11 (2015) 1655–1658.
[10] J.F. Romero-Arauz, C. Carranco-Salinas, A. Leaños Miranda, O.A. Martínez-Rodríguez, Preeclampsia atípica. Reporte de un caso, Ginecol. Obstet. Mex. 82 (2014) 354–360.
[11] A.L.Tranquilli G., Dekker L. Magee, The classification, diagnosis and management of the hypertensive disorders of pregnancy: a revised statement from the isshp, Preg. Hypertens. 4 (2) (2014) 97–104.
[12] B.M. Sibai, C.L. Stella, Diagnosis and management of atypical preeclampsia-eclampsia, Am. J. Obstet. Gynecol. 200 (5) (2009) 481–482.
[13] L.C. Poon, I. Staboulidou, N. Maiz, W. Plasencia, K.H. Nicolaides, Hypertensive disorders in pregnancy: screening by uterine artery Doppler at 11–13 weeks, Ultrasound Obstet. Gynecol. 34 (2) (2009 Aug) 142–148.
[14] L.C. Poon, N. Maiz, C. Valencia, W. Plasencia, K.H. Nicolaides, First-trimester maternal serum pregnancy-associated plasma protein-a and pre- eclampsia, Ultrasound Obstet. Gynecol. 33 (1) (2009 Jan) 23–33.
[15] K. Spencer, N.J. Cowans, I. Chefetz, J. Tal, H. Meiri, First-trimester maternal serum PP-13, PAPP-A and second-trimester uterine artery doppler pulsatility index as markers of pre-eclampsia, Ultrasound Obstet. Gynecol. 29 (2) (2007 Feb) 128–134.
[16] W. Plasencia, N. Maiz, S. Bonino, C. Kaihura, K.H. Nicolaides, Uterine artery doppler at 11 + 0 to 13 + 6 weeks in the prediction of pre-eclampsia, Ultrasound Obstet. Gynecol. 30 (5) (2007 Oct) 742–749.
[17] L.C. Poon, I. Staboulidou, N. Maiz, W. Plasencia, K.H. Nicolaides, Hypertensive disorders in pregnancy: screening by uterine artery Doppler at 11–13 weeks, Ultrasound Obstet. Gynecol. 34 (2) (2009 Aug) 142–148.